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Obstructive choledocholithiasis requiring intervention in a three week old neonate: A case report and review of the literature



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ABSTRACT

The discovery of cholelithiasis in neonates is often incidental, however obstructing common bile duct stones are rare. Herein we report the case of a 3 week old neonate who presented with obstructive choledocholithiasis. The patient was treated conservatively with antibiotics and ursodeoxycholic acid but did not improve. He was therefore taken to surgery for cholecystectomy and stone extraction. The operation was successful and his transaminases and bilirubin levels declined. Trials of conservative management can be attempted in asymptomatic infants with choledocholithiasis. However, failure of the stone to pass or ongoing signs of cholecystitis should be met with operative intervention to remove the obstruction.

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Cholelithiasis is often an incidental finding in the neonatal population. Obstructing common bile duct stones, however, are quite rare. Herein we report the case of a 3 week old neonate with choledocholithiasis and acute cholecystitis who required operative intervention to relieve the obstructing stone.

1. Case report

The patient is a 3 week old, 3.9 kg infant male born at 39 weeks and 3 days gestation. He is blood type B+, coombs negative, and had a normal newborn screen. He was referred to the hospital with a working diagnosis of pyloric stenosis, as he was experiencing projectile emesis and failure to thrive. However, blood chemistries revealed elevated transaminases (ALT 166 Units/L, AST 324 Units/L) and bilirubin (total bilirubin 2.7 mg/dL, direct bilirubin 0.6 mg/dL). Abdominal ultrasound showed gallbladder sludge, a cystic duct stone, pericholecystic fluid, and a normal pylorus (Fig. 1).

He was admitted for fluid resuscitation and was started on cefepime. Laboratory assessment the following day showed worsening bilirubin values (Fig. 2), and a repeat ultrasound showed that the stone was now lodged in the common bile duct near the pancreatic head. The common bile duct was dilated proximally to

4–5 mm and the stone measured $4 \times 7 \times 3$ mm. He was started on ursodeoxycholic acid and surgical consultation was obtained. As he was virtually asymptomatic, he had serial daily ultrasounds and blood chemistry assessments for 3 days to determine if the stone would pass spontaneously. Assessment of these parameters did not indicate resolution of the stone. He then underwent MRCP (Fig. 3) to assess his biliary anatomy and to ensure that there was no choledochal cyst. The stone was confirmed to be lodged in the common bile duct at the pancreatic head, the common bile duct was dilated to 4–5 mm, and there was no anatomic biliary ductal abnormality appreciated. As the stone was not passing and there were continued signs of cholecystitis on imaging, we proceeded to the operating room for cholecystectomy and intraoperative stone extraction. ERCP was not an option as our gastroenterologists did not have an endoscope small enough to complete the procedure.

In the operating room, a right subcostal incision was made and the gallbladder was readily identified under the liver edge. It was fairly hard in texture. A cholangiogram was performed and clear bile was expressed from the gallbladder (Fig. 4A). The cholangiogram noted patent right and left hepatic ductal systems and an obstructive stone at the distal common bile duct (Fig. 4B). The biliary system was flushed several times with normal saline but this did not move the stone. We then proceeded to pass a 2 French Fogarty catheter through the gallbladder and into the distal biliary system but the catheter was too pliable and it created a false passage in the gallbladder lumen. We then opened the gallbladder

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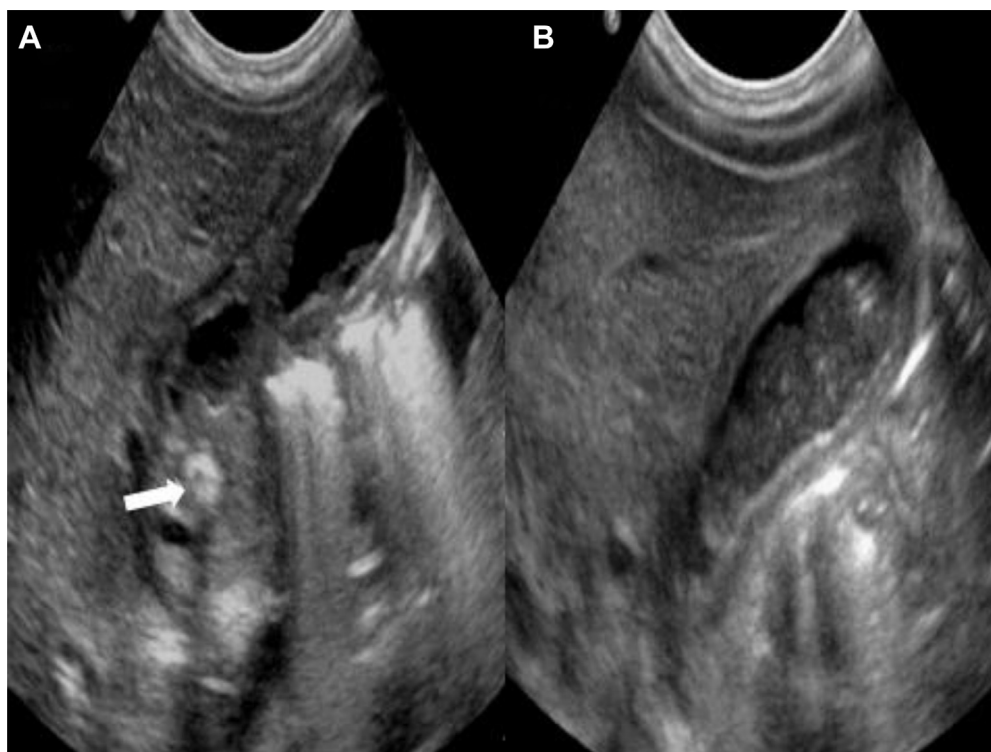


Fig. 1. (A) Abdominal ultrasound denoting the longitudinal view of the gallbladder with a visualized calculus in the cystic duct (white arrow). (B) Longitudinal view of the gallbladder showing sludge within the lumen.

lumen longitudinally toward the cystic duct orifice. A 0.035" Glidewire (Boston Scientific) was then passed into the distal bile duct and a 3 French open ended ureteral stent catheter (Cook) was deployed over the wire. The Glidewire was then withdrawn and a 1.5 French stone extraction basket (Cook) was deployed through the ureteral stent. It was able to pass around the stone, and a portion of the stone was retrieved (Fig. 5A). A subsequent cholangiogram demonstrated a portion of the stone still in place (Fig. 5B). A second

pass of the basket was performed and this action dislodged the stone into the duodenum thereby alleviating the obstruction and allowing free flow of contrast into the duodenum. The ureteral stent and basket were then removed and a cholecystectomy was performed.

The day after the surgery his bilirubin was still elevated to a total of 3.0 mg/dL but this dropped to 2.1 mg/dL with a direct component of 1.3 mg/dL. He was discharged from the hospital with instructions to follow up in clinic in two days. He came back to clinic on post-operative day 6 and his total bilirubin had normalized to 1.0 mg/dL and his direct component was just slightly elevated at 0.4 mg/dL. He was eating and stooling normally and gaining weight as expected. At post-operative day 21, his laboratory parameters had normalized.

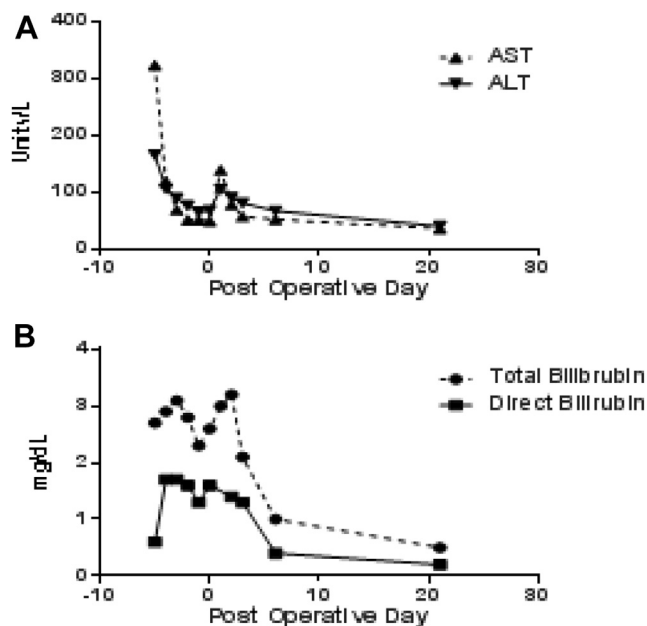


Fig. 2. Progression of (A) liver transaminases and (B) bilirubin levels over the course of hospitalization (AST and ALT in Units/L, Total bilirubin and Direct Bilirubin in mg/dL).



Fig. 3. Magnetic resonance cholangiopancreatography image demonstrating a dilated common bile duct and obstructing common bile duct stone (white arrow).

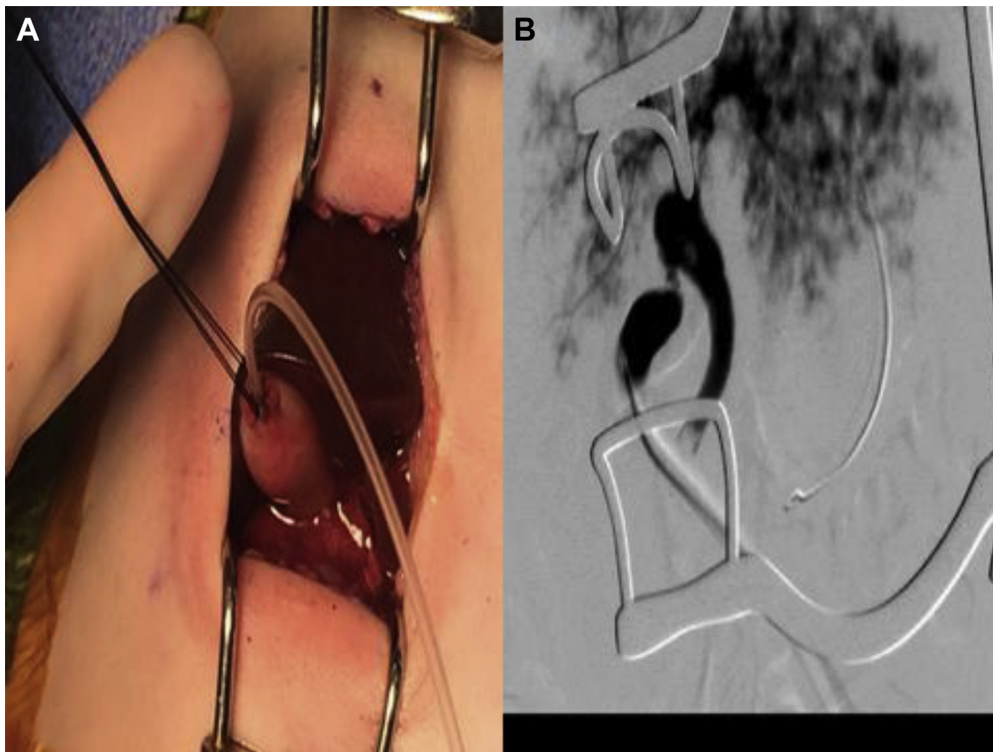


Fig. 4. (A) Intraoperative photograph of cholangiogram catheter in fundus of gallbladder. (B) Initial cholangiogram depicting patent right and left hepatic ducts and obstructive stone in distal common bile duct with no passage of contrast into the duodenum.

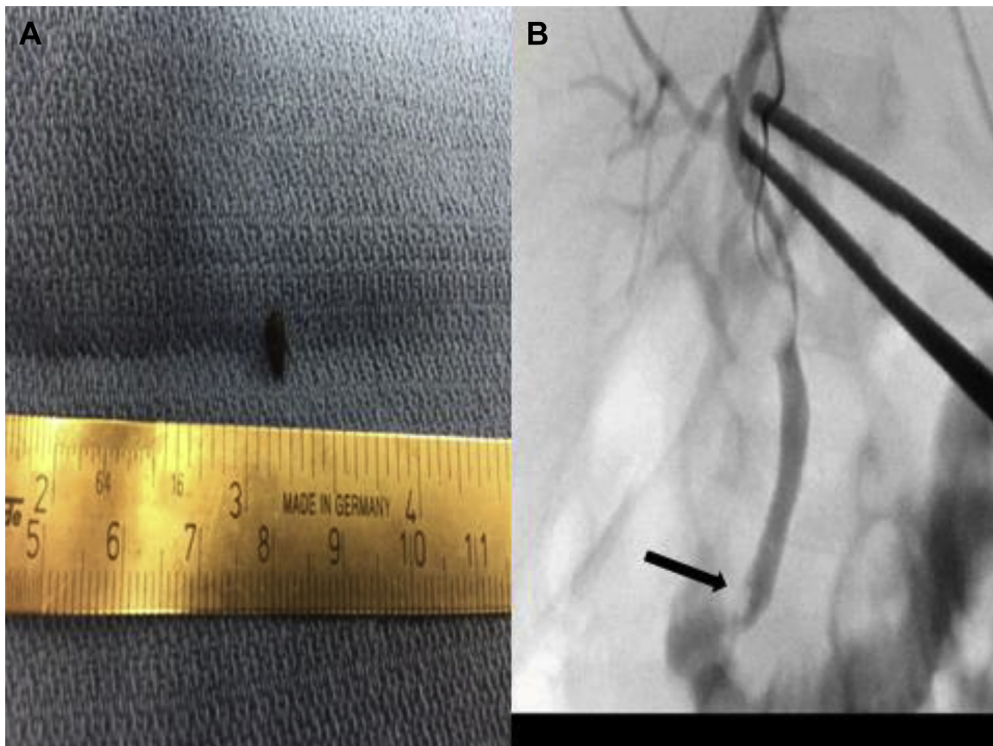


Fig. 5. (A) Portion of common bile duct stone retrieved by stone basket. (B) Repeat intraoperative cholangiogram demonstrating partial removal of stone. Black arrow indicates residual stone.

2. Discussion

Biliary sludge and cholelithiasis are not uncommon in the neonatal and early childhood periods. Documented risk factors for developing cholelithiasis include dehydration, hemolysis, urinary tract infection, cholestatic liver disease, total parenteral nutrition, immunoglobulin A deficiency, prematurity, and certain metabolic derangements such as glucose-6-phosphate deficiency [1,2]. Many times, cases are incidentally found and resolve spontaneously without the need for intervention.

This patient had no evidence of dehydration on clinical examination or chemistry studies. He had normal hemoglobin and reticulocyte counts which suggested that red blood cells were not being hemolyzed. An infectious etiology was originally considered, but his blood and urine cultures were normal and he did not exhibit a leukocytosis. He was not on parenteral nutrition and had no family history or previous personal history of disorders of metabolism. We therefore concluded that his stones were idiopathic in nature.

The literature is fairly lacking in the documentation of choledocholithiasis in the neonatal population, although a few small case series do exist. Conservative therapy appears to be the mainstay, with operative intervention reserved for patients in whom the stones do not pass or cholecystitis persists. A 2012 case series documented three patients with obstructive choledocholithiasis and associated transaminitis. All three patients responded to treatment with ursodeoxycholic acid and antibiotics within 48 h of therapy, and all three had resolution of the stone by ultrasound criteria 11 days following diagnosis. Authors suspected that the antibiotics helped to reduce inflammation within the biliary tree and the ursodeoxycholic acid helped to improve bile flow [3]. Two other case reports documented successful conservative therapy for infantile choledocholithiasis. One report noted choledocholithiasis in a 5 month old former premature infant who was having persistent vomiting. This child was also started on antibiotics and ursodeoxycholic acid [4]. The other neonate had no risk factors and had a common bile duct stone noted on ultrasound [5]. Both children's stones resolved spontaneously.

A case series published in 1990 reported on 7 infants with choledocholithiasis. Two patients recovered spontaneously and the remaining five required intervention via surgery or endoscopic therapies [6]. Although many patients seem to have spontaneous resolution of stones, there are several reports of interventions being required to alleviate the obstruction. An earlier study in 1985 documented two infants under 4 months of age who both required laparotomy and common bile duct exploration for stone extraction [7]. A more recent 2005 report notes the successful treatment of a 2 month old infant with choledocholithiasis with open cholecystectomy and T tube drainage [8], while another case utilized a cholecystostomy tube to irrigate the biliary system and dislodge the stone [9]. Other cases have demonstrated increased abdominal distention and ascites associated with choledocholithiasis and common bile duct rupture which required operative intervention [10].

The use of ERCP has been sparsely documented in the neonatal period. Two case reports document the successful treatments of a 4 month old and a 6 month old with ERCP and sphincterotomy [11,12]. Another report notes an 8 month old with an obstructive stone who was treated with antibiotics for two weeks followed by ERCP at 9–10 months of age and laparoscopic cholecystectomy another month following endoscopic intervention [13]. Although ERCP is not routinely utilized in newborn infants due to their small size, centers with infant ERCP capabilities are encouraged to use this modality as the initial invasive procedure to clear the duct and allow for resolution of the inflammatory process.

There are currently no guidelines for the timing or method of relieving the obstructive stone in the early infant population. Watchful waiting with antibiotics and ursodeoxycholic acid is appropriate as long as symptoms of cholangitis are not present. However, if the stone does not resolve or symptoms of cholangitis ensue, operative intervention is required. ERCP should be considered first line therapy if experienced personnel are present with optimal equipment. Radiographic placement of a percutaneous cholecystostomy tube and initiation of common bile duct flushing may be feasible if patients are too sick for operative intervention. However, their use requires leaving a tube in place for several weeks after the procedure, which may not be an attractive option for families.

Ultimately, operative ductal exploration and cholecystectomy may be necessary if preoperative imaging denotes a choledochal cyst or if the stone fails to pass. Open or laparoscopic approaches are possible, and should be left up to the surgeon based on skill levels, keeping in mind that a laparoscopic approach may only be feasible if duct exploration is not required (i.e. ERCP performed first to clear the duct). Duodenotomy and transduodenal approaches should only be utilized if transductal approaches fail to relieve the obstruction due to complications associated with duodenal leak and potential injury to pancreatic and biliary ductal structures. Following duct exploration, a cholecystectomy should be performed so as to eliminate future sludge and stone formation.

3. Conclusion

Cholelithiasis in the neonatal period is not uncommon, but choledocholithiasis is fairly rare. Many reports have documented the successful resolution of obstructive stones with antibiotics and ursodeoxycholic acid. Nonetheless, in instances where passage of the stone is not progressing or infants have continued signs of cholecystitis on laboratory values or imaging despite medical therapy, then surgical intervention should be considered. Clinicians are encouraged to balance costs and risks of watchful waiting with operative intervention.

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